Clinical note

SPECT-CT in the localization of an ectopic retropharyngeal parathyroid adenoma as a cause for persistent primary hyperparathyroidism

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ABSTRACT

A common cause for surgical failure of primary hyperparathyroidism (pHPT) is the non-detection of an ectopic adenoma during the intervention. We present a case of a patient with pHPT in whom an ectopic gland was found in the right retropharyngeal space by means of a double phase 99mTc-methoxy-isobutylisonitrile (99mTc-MIBI) scintigraphy and early SPECT-CT after several surgeries and imaging tests.

Adding a tomography to the planar scintigraphy increases its sensitivity and improves pathological parathyroid glands localization. The hybrid imaging is sometimes essential to obtain surgical success, as in the case of ectopic adenomas.

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Valor del SPECT-TAC en la localización de un adenoma de paratiroides ectópico retrofaringeo como causa de hiperparatiroidismo primario persistente

Palabras clave:
Adenoma ectópico
Gammagrafía de paratiroides con 99mTc-MIBI
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R E S U M E N

Una causa frecuente de fracaso de la cirugía del hiperparatiroidismo primario (HPTp) es la existencia de un adenoma ectópico no detectado en la intervención. Presentamos a una paciente con HPTp en la que, tras sucesivas intervenciones y pruebas de imagen, se encuentra una glándula ectópica en el espacio retrofaringeo derecho mediante gammagrafía de doble fase con 99mTc-metoxi-isobutil-isonitrilo (99mTc-MIBI) y SPECT-TAC precoz.

La adición de una tomografía al estudio gammagráfico planar, aumenta su sensibilidad y mejora la localización de las paratiroides patológicas. En ciertas ocasiones, como es el caso de adenomas ectópicos, la imagen híbrida será esencial para conseguir el éxito quirúrgico.

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Introduction

The most frequent causes of persistence of primary hyperparathyroidism (pHPT) after surgery are the absence of identification of an adenoma (mainly due to ectopic parathyroid glands), the presence of unsuspected multilobular disease (MGD) and insufficient extension of the surgery in known MGD.1

Double-phase scintigraphy with 99mTc-methoxyisobutylisonitrile (99mTc-MIBI) is a diagnostic tool of demonstrated utility in the detection of pathologic parathyroid glands prior to surgery, especially in cases of ectopic localizations.2 Nonetheless, it should not be forgotten that the sensitivity diminishes in cases of MGD.3

The addition of SPECT to conventional studies in the last years has notably increased the sensitivity of scintigraphy,4 in both double-phase and double-tracer studies, and it has been described that SPECT-CT particularly improves the localization of ectopic parathyroid glands.5 Likewise, the combination of scintigraphy in any of its protocols with morphological imaging methods, mainly ultrasound,6 increases the diagnostic precision. CT or MR studies are generally reserved for cases of failure to detect the pathological parathyroid gland by other imaging methods or in the presence of ectopic parathyroid glands with the aim of providing an anatomical substrate for the planning of the surgical approach.7

Clinical case

A 61-year-old woman diagnosed with persistent pHPT was referred to the Department of Nuclear Medicine for a parathyroid SPECT-CT scan with 99mTc-MIBI. With regard to her clinical history, she had undergone surgery on 4 occasions. The first intervention was in 1998 [previous serum calcium values between 10.5 mg/dl and 11 mg/dl; range of normality: 8.5–10.4 mg/dl, and intact parahormone (I-PTH) was 153 pg/ml; range of normality:
11.1–79.5 pg/ml] and involved extirpation of the 2 upper parathyroid glands and the left inferior gland, the latter being pathologic. The low right parathyroid gland was not identified. During the immediate post-operative period, a transitory reduction of I-PTH was observed but without complete normalization. Different studies were therefore requested including double-phase scintigraphy with $^{99m}$Tc-MIBI, cervical ultrasound, cervical-thoracic MR and supraaortic trunk angiography with the aim of localization, albeit unsuccessfully. In addition, selective venous parathyroid catheterism was performed obtaining I-PTH samples showing hormone hypersecretion (I-PTH = 946 pg/ml) in the sample from the right internal jugular vein. In view of this result, a second intervention involving right hemithyroidectomy was carried out in 2001 in which no pathological findings were detected (post-operative serum calcium levels = 11.6 mg/dl and I-PTH = 173 pg/ml).

Posteriorly, on suspicion of left parathyroid adenoma described in a double-phase scintigraphic study with $^{99m}$Tc-MIBI, another intervention was performed in 2002, with no pathological parathyroid evidence (post-surgical serum calcium levels = 10.6 mg/dl and I-PTH = 276.6 pg/ml).

From 2002 to 2008 multiple studies were undertaken: double-phase scintigraphy with $^{99m}$Tc-MIBI, cervical ultrasound, cervical-thoracic CT and MR and $^{11}$C-methionine PET, with no significant findings. Likewise, another selective venous catheterism was performed obtaining samples for I-PTH and showing hormone hypersecretion at a cervical level with right lateralization. With these results, a new intervention was planned in the same year, completing the thyroidectomy. The I-PTH values persisted at 208.77 pg/ml following surgery. In January 2011 the I-PTH rose to 481 pg/ml and the patient also presented numerous episodes of nephritic colic due to renal lithiasis. A double-phase scintigraphy was performed next to the right submaxillary gland, being inferior-medial and slightly posterior to the gland. This uptake was of little relevance in the early planar study (10 min post-injection) due to the proximity to the submaxillary gland and the intensity of the uptake and also showed rapid washout, disappearing in the delayed image (2 h post-injection). This activity, which may be due to an ectopic parathyroid gland, was localized in the fused images in the right retropharyngeal space at the level of the thyroid cartilage.

With these results, another surgical intervention was performed, localizing a parathyroid adenoma with a weight of 59.7 mg in the region described in the scintigraphic study. Both the serum calcium and I-PTH values are currently within the normal range (I-PTH = 59.9 pg/ml and calcium = 9.1 mg/dl).

**Discussion**

During their embryonary development the superior parathyroid glands originate in the 4th branchial arch and the inferior glands and the thymus originate in the 3rd branchial arch.\(^8\) It is therefore possible to find inferior parathyroid glands in the interior of the thymus and even in the anterior mediastinum, while the superior glands may emigrate toward the posterior mediastinum. The most frequent perithyroid ectopias are lateral (near or in the carotid sheath), thymothyroid ligament (in a low position), and inter-cricothyroid, retropharyngeal and intra-thyroid. The most frequent distant ectopias are thoracic (anterior or, more rarely, middle mediastinum) and undescended parathyroid glands at the level of the carotid bifurcation.\(^1\) In our case, the ectopic parathyroid gland, undescended, inferior right parathyroid gland, was found in a retropharyngeal submandibular location.

Imaging studies are essential for planning a minimally invasive surgical approach. They are also recommended in cases of bilateral surgery in patients with previous cervical surgery hindering surgical examination, and after failure in a previous intervention to localize other pathologic parathyroid glands in MGD or ectopic parathyroid glands. Nonetheless, depending on the localization, the ectopic gland may be difficult to detect. In morphological imaging studies, an ectopic submandibular localization, as in the present case, may be mistaken with adenopathies, which are frequent in the cervical region. Functional images may not adequately differentiate the parathyroids when only a planar image is made since the parathyroid glands may be masked by some anterior structure or any proximal structure with an equal or more intense uptake similar to what occurred in our patient.\(^8\) As shown, SPECT is a helpful tool in these situations since it improves spatial resolution. In addition, it has been described that the localization of parathyroid adenomas, especially ectopic cases,\(^5,10\) notably improves with the use of hybrid equipment.

In the interpretation of the studies, as in our case, it should be taken into account that in the presence of rapid washout adenomas conventional double-phase scintigraphy is of less aid, and the addition of early SPECT increases the diagnostic safety.\(^11\)
Fig. 2. Early SPECT-CT (axial, sagittal and coronal slices): fusion images (A), SPECT (B) and CT (C). The pathological parathyroid gland is localized in the right retropharyngeal space at the level of the thyroid cartilage.

References